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Urinary Obstruction Due to Fungal Infection in Two Preterm Siblings

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Authors' contributions

This work was carried out in collaboration between all authors. Authors VLD, CMP and ALS wrote the draft of the manuscript. Authors RS, CMS and TB managed the literature searches. Author SC designed the figures, managed literature searches and contributed to the correction of the draft. Authors MB, AM, ESB and OM provided the case, the figures and supervised the work. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Extravasation of urine and formation of urinoma is rare in the absence of either trauma or congenital anomalies. Fungal ball formation in urinary tract can cause obstruction leading to extravasation. We present two such cases in siblings. *Case One*: Triplet II referred from the Neonatal unit with clinical picture of acute abdomen. Imaging was inconclusive therefore he

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proceeded to an exploratory laparotomy, where a diagnosis of urinary ascites was made. *Candida albicans* infection was confirmed on urine culture. He was treated with antifungals and made a full recovery. No structural anomalies were demonstrated on renal tract imaging. *Case Two*: Triplet I presented to ED one month later with symptoms of respiratory infection. Commenced on empirical antibiotic medications but clinically deteriorated. Urine culture was positive for fungal infection so antifungal medication was commenced as well. USS demonstrated a right perinephric urinoma. This was drained percutaneously. He made a good recovery and no structural anomalies were demonstrated on renal tract imaging. Although there is no other direct link between the two cases, that the patients are siblings may suggest a congenital factor. Anyhow, early recognition is vital in managing these complicated patients. Further study is required to explore the concept of a potential familial predisposition for the condition.

Keywords: Candidiasis; neonate; urinary obstruction; urinoma.

1. INTRODUCTION

Extravasation of urine is most commonly associated with traumatic injury to the urinary tract and often leads to the formation of urinoma or urinary ascites. Most cases are associated with congenital obstruction of urinary flow secondary to either posterior urethral valves or pelviureteric junction obstruction [1-4]. In these patients the extravasation is due to increased pressure within the urinary system and this can be associated with a rupture. In very rare cases this obstruction, which leads to extravasation, is not due to a congenital anomaly, but to the presence of fungal balls forming within the urinary tract [5,6]. Although rare, this has been demonstrated in preterm neonates receiving long term antibiotic treatment who have developed a systemic fungal infection [5,6]. Early diagnosis and appropriate treatment are essential to effectively manage this potentially damaging complication. We report two interesting cases of neonatal urinary tract obstruction secondary to candidiasis in triplets.

2. CASE PRESENTATION

2.1 Case One

The male infant in the first case was born in a Level 1 Neonatal Unit t 36 weeks with a birth weight of 1250g and was the second of triplets. He initially made routine progress, establishing enteral feeds and growing steadily. He had several episodes of suspected sepsis and therefore had prolonged exposure to antimicrobials, without antifungal cover. On day 54 of life he suffered a clinical deterioration associated with decreased enteral feed tolerance, increased aspirates and abdominal distension. Despite conservative management including bowel rest, the abdominal distension remained and his bowels had not opened for three days. With the clinical picture worsening, he was referred to the Surgical department on the third day with suspected abdominal pathology.

On examination the abdomen was distended and hard, non-tender with no skin changes, but an oedematous abdominal wall (associated weight gain of 500 g over three days). Blood results from the time demonstrated an inflammatory process (raised WCC and CRP, dropping platelet count). Abdominal x-ray showed dilated loops of bowel, with no obvious free air. Ultrasonography (USS) revealed free fluid within the peritoneal cavity and in the retroperitoneal space, bilateral hydronephrosis and pericardial and pleural effusions.

Due to the unknown aetiology a midline exploratory laparotomy was performed. Intraoperatively free clear, yellow fluid was found within the peritoneal cavity and similarly in the retroperitoneal space, once opened. An ascitic fluid specimen was sent to biochemistry and microbiology. Bilateral renal capsules were intact but grossly hydronephrotic. The bowel loops were examined but no pathology was identified. A peritoneal drain and urethral catheter were inserted and the cavity closed.

The ascitic fluid was confirmed to be urine and both the bladder and peritoneal specimens cultured Candida albicans. This was treated with caspofungin as per the local microbiology advice (eleven days total). He continued to receive the full support from the Neonatal unit and made good progress post-operatively. The drain and catheter were removed after ten and eleven days respectively. Post-operative imaging, including a micturating cystourethrogram and renal MRI, failed to demonstrate any structural anomalies within the renal tract. Serial USSs were performed demonstrating a steady reduction in

residual urinoma, with complete resolution by six weeks. He was transferred to the local Neonatal unit on post-operative day twelve and discharged home after a further two months. He continues to make good progress.

2.2 Case Two

The second case presented a month later and was the sibling triplet of the first case. Born at 36 weeks he was Triplet I and his birth weight was 1230 g. He made steady progress during his three month stay on the Neonatal unit. His only significant problems were multiple courses of various antibiotics for episodes of suspected sepsis (lower respiratory tract infections. He was discharged home at the corrected gestational age of 51 weeks and weight of 3700 g.

He presented to Paediatric Emergency department (PED) five days after discharge with decreased oral intake and episodes of pyrexia. Bloods tests showed an inflammatory process (raised WCC and CRP) and anaemia, but otherwise normal biochemistry. Chest x-ray confirmed the clinical suspicion of pneumonia

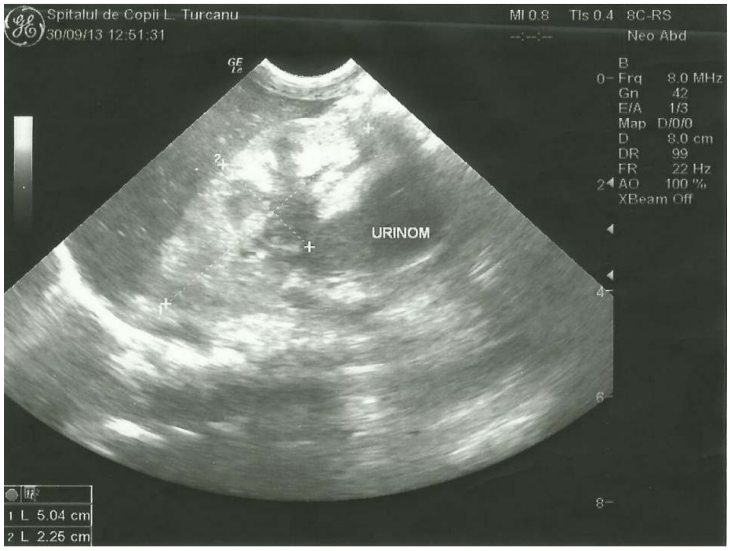

Fig. 1. Residual urinoma on the right side 18 days after surgery - Case 1

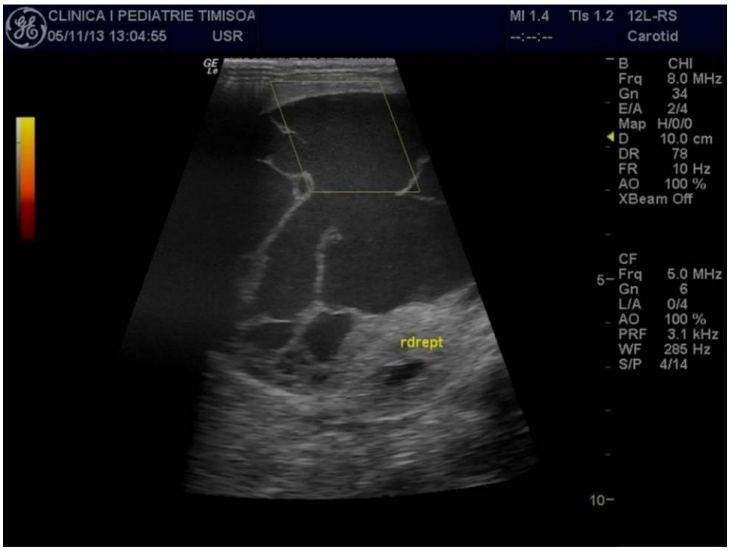

Fig. 2. Right perirenal fluid accumulation with mass effect on the right kidney, unomogenous content and no Doppler flux - Case 2

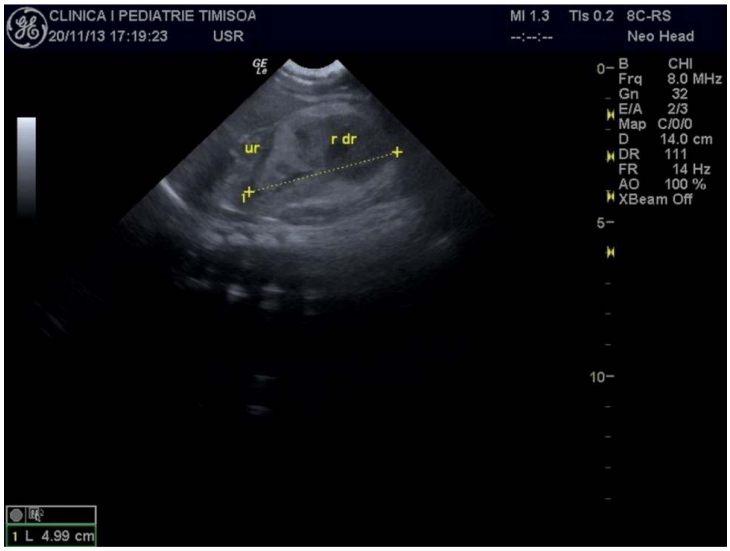

Fig. 3. Fifteen days from surgery the perirenal fluid accumulation disappeared - Case 2

and empirical antibiotic treatment was commenced. He was admitted under the Paediatric team for further management. USS of the renal tract suggested inflammation of the right kidney with poor corticomedullary differentiation. An immediate urine microscopy and culture showed fungal growth therefore he was started on empirical antifungals (fluconazole) for possible fungal pyelonephritis. Over the following five days he developed abdominal distension, constipation and a palpable mass in the right flank. USS at the time revealed large (6x8 cm) perinephric cystic lesion compressing the right kidney. There was no free fluid seen within the peritoneal cavity. Triplet I was referred to the Surgical team and a percutaneous drain was inserted into the collection under ultrasonographic guidance. A sample of drain fluid was sent for analysis and confirmed to be *Candida albicans*. The treatment was changed appropriately, to caspofungin and was continued for the next 11 days. He made a good recovery post-intervention and further imaging demonstrated a good resolution of the urinoma. The drain was removed on day eight after the procedure and patient was discharged home on day 15 post-procedure. He, too, continues to progress well since discharge.

3. DISCUSSION

Candidiasis is a well recognised complication of long-term antibiotic treatment. In preterm babies this has severe implications; it is more difficult to recognise and has a poorer prognostic outcome than in other age groups. Systemic fungal infection can involve formation of fungal balls within the renal tract. In very rare cases these can migrate to the physiological narrow portions of the urinary system and cause acute obstruction [5-7], dilatation of the upper tract and subsequent renal failure. If left untreated, this can lead to urinary tract rupture and formation of urinoma.

Unfortunately this occurred in both the cases presented. In the first there was bilateral obstruction, followed by rupture and formation of a retroperitoneal urinoma. This mimicked the symptoms of peritonitis and resulted in an exploratory laparotomy. Ideally percutaneous nephrostomy is the salvage procedure in these cases with no further interventions required [7-9]. Fortunately the lessons were learnt from the first case and when his sibling presented the diagnosis was considered, recognised early and a simple, minimally invasive procedure was successfully performed. After appropriate drainage and antifungal treatment both patients made a good recovery and no other complications were noted.

Perhaps the most interesting feature in these cases is that the two patients are siblings. Although there is no other direct link between the two cases, that the patients are siblings may suggest a congenital factor. Clinical examination and radiological investigations have not demonstrated any abnormalities of the urinary tract. We postulate that, in association with the fungal infection and the low birth weight, there is a familial physiological predisposition for

obstruction within the urinary system. This is supported by the fact that the second case developed urinary obstruction despite commencing antifungal treatment.

Early recognition of the condition is vital, but unfortunately this is difficult due to the rarity of its presentation [5-9]. Delays in diagnosis and initiation of appropriate treatment can cause increased morbidity. An example is the first case, where a potentially avoidable laparotomy was carried out. Therefore it is crucial for all of the Multidisciplinary team, including Radiology to be aware of this diagnosis and consider it in a similar future situation.

4. CONCLUSIONS

Fungal infections are a major cause of morbidity, even mortality, in premature babies, despite appropriate prophylaxis. The key to timely management and treatment is early recognition and diagnosis. Although urinoma secondary to fungal ball obstruction is a rare finding it should be considered in cases with similar presentation. Further study is required to explore the concept of a potential familial predisposition for the condition.

CONSENT

All authors declare that written informed consent was obtained from the parents of the patients for publication of this case report and accompanying images.

ETHICAL APPROVAL

All authors hereby declare that all experiments have been examined and approved by the appropriate ethics committee of the hospital and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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